DOES COT DEATH STILL EXIST?

With leading researchers saying smoking and other modifiable factors account for most sudden infant deaths, **Jonathan Gornall** asks whether it is time to put the diagnosis to bed

There are no cases of sudden infant death syndrome (SIDS) in Vanderburgh County, Indiana. "I simply don't believe in SIDS deaths," says chief deputy coroner Annie Groves. "If you do a complete investigation, you will find a cause of death."

Coroner Groves's confidence may sit uncomfortably with many professionals in the United Kingdom, where even the use of the word "unascertained" in unexplained infant deaths is frowned upon because it carries "implications that the death may have been the result of neglect or abuse."[1] But in America she is not alone in her certainty.

The Vanderburgh experience is one of many highlighted by a review of 40 000 infant deaths between 1992 and 2004 that concludes the quality of investigations carried out into sudden infant deaths in America "varies irrationally."[2]

The nationwide review, conducted by the Scripps Howard News Service, found that the protocol for the investigation of sudden unexpected deaths in infancy launched by the Centers for Disease Control and Prevention in 2006 was mandatory in only seven states. In those areas where the protocol was applied, far fewer deaths were ascribed to SIDS and medical examiners and coroners were more willing to conclude that inappropriate parenting was the cause of sudden unexpected deaths in infancy.

"It is far more common for a child to die of [unintentional] asphyxiation than to die from SIDS," Andrea Minyard, the state medical examiner in Pensacola, Florida, told the survey. "We say this with a heavy heart. But it is an accurate portrayal of what really is happening."

Vanishing diagnosis

The term SIDS was introduced in 1969,[3] "partly for humanitarian reasons, being intended as a recognized category of natural death that carried no implication of blame for bereaved parents."[4] Since then, however, a lot has been learnt about the major modifiable risk factors and the role of parenting in cot death. The number of cases in the UK has fallen by 75% since the 1991 Back to Sleep campaign.[5]

In 2005 there were 2107 deaths registered as SIDS in America[6] and 191 in England and Wales,[7] but from this April all sudden unexpected infant deaths in the UK will be investigated according to a new national multiagency protocol that is expected to reduce by half the number of deaths registered as SIDS.[8]

Meanwhile, in October Britain's leading SIDS research team concluded that maternal smoking during pregnancy—already a recognised factor in 90% of cot death cases—met the criteria for causality and was directly responsible for 60% of such deaths.[9]

The Foundation for the Study of Infant Death, which funds the work of the Bristol research unit, endorsed this conclusion. George Haycock, the foundation's scientific adviser, delivered this plain message: "If no women smoked in pregnancy, about 60% of cot deaths could be avoided. This means that in the UK the number of deaths could fall from around 300 a year to 120 a year."[10]

It seems that SIDS, a spectre that for more than 40 years has caused fear and anxiety for countless parents, is simply withering away in the face of closer and closer scrutiny, undermining the popular myth propagated by much media coverage that cot death is a bolt from the blue that can strike any child from any family.

Smoking is, of course, not the only advice being missed or ignored by the majority of parents whose children die from SIDS, as the American experience chronicled by Scripps Howard anecdotally attests.

In a 2006 paper, Peter Fleming and colleagues at Bristol described how the epidemiological profile of SIDS had changed between 1984 and 2003, partly because of the impact of the Back to Sleep campaign. The most worrying finding was the increase from 57% to 86% in the proportion of mothers who smoked during pregnancy, but also of concern was the enduring prevalence of inappropriate sleeping position.

Before the campaign, 89% of babies dying from SIDS in Avon had been put down to sleep on their fronts. Although by 2003 there had been a large reduction, babies who had been placed to sleep in the prone position still accounted for a quarter of SIDS deaths.

The proportion of SIDS babies who died while co-sleeping with their parents had risen from 12% to 50% of cases. The number of deaths in the parental bed had halved but the number of co-sleeping deaths on sofas had risen.[5]

Tony Risdon, the only forensic paediatric pathologist in the UK, is based at Great Ormond Street Hospital, where his department sees about one third of all infants who have died suddenly and unexpectedly. He never uses the term SIDS: "When you get down to it, the cases that absolutely fulfil all the international criteria of SIDS are a tiny minority and if every parent followed

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Centers for Disease Control and Prevention protocol for sudden unexpected infant deaths

In March 2006 the CDC released an eight page questionnaire based reporting form designed to elicit essential basic information in the investigation of all sudden unexpected infant deaths (www.cdc.gov/SIDS/SUID.htm).

The form is designed to be used by whoever interviews the witness (coroner, death scene investigator, police officer, or medical examiner). It includes 79 questions divided into five categories:

- Witness interview
- Infant medical history
- Infant dietary history
- Pregnancy history
- Incident scene investigation.

A section for investigator's notes includes templates for diagrams of the scene of death and observed marks or injuries on the body.
The form concludes with a summary for the pathologist to indicate whether preliminary investigation suggests any of the following:

- Asphyxia (overlying, wedging, choking, nose/mouth obstruction, rebreathing, neck compression, immersion in water)
- Sharing of sleeping surface with adults, children, or pets
- Change in sleeping condition (unaccustomed stomach sleep position, location, or sleep surface)
- Hyperthermia/hypothermia (excessive wrapping, blankets, clothing, or hot or cold environments)
- Environmental hazards (carbon monoxide, noxious gases, chemicals, drugs, devices)
- Unsafe sleeping conditions (couch/sofa, waterbed, stuffed toys, pillows, soft bedding)
- Diet (introduction of solids, etc)
- Recent hospital admission
- Previous medical diagnosis
- History of acute life threatening events (apnoea, seizures, etc)
- History of medical care without diagnosis
- Recent fall or other injury
- History of religious, cultural, or ethnic remedies
- Death due to natural causes other than SIDS (birth defects, complications of preterm birth)
- Prior sibling deaths
- Previous encounters with police or social service agencies
- Request for tissue or organ donation
- Objection to autopsy
- Pre-terminal resuscitative treatment
- Death from (injury), poisoning, or intoxication
- Suspicious circumstances.

the advice on safe sleeping environments and so on, this problem would probably disappear."

Thanks to ongoing controversy over how some sudden infant deaths are categorised, even the scale of the problem in the UK remains uncertain.[11 12] The Foundation for Study of Infant Deaths, Britain's leading funder of cot death research, says that in 2005 there were 300 cot deaths in the UK,[13] but this figure includes 77 deaths registered in England and Wales as "unascertained."[7]

Time for reassessment

However many SIDS cases there are, the fact that by the foundation's own estimation most are now attributable to modifiable parental behaviour suggests the need for a fundamental rethink. For a start, given that the definition of SIDS includes the requirement that the death remains unexplained, it seems illogical to continue to classify as SIDS those deaths to which a clear cause has been attributed.

Furthermore, does it make sense to continue investing scarce research funds in the search for a "cause" for the decreasing number of unexplained infant deaths? Many talented scientists have spent entire careers working to identify the cause, or causes, of SIDS. Beyond the elimination of suspects, however, most have got nowhere. As Abraham Bergman, one of the medical founding fathers of the SIDS movement, commented in 1997, "It is my subjective impression that over 80% of published papers about SIDS contain conclusions that have not been substantiated."[14]

After 40 years, scientists can't even agree on where to look for an answer. A genetic susceptibility is considered the most likely source of the problem by some,[15] while others are equally adamant that SIDS is a developmental disorder.[16]

Most, however, do agree that a proportion of babies are either born with or contract one or more largely unidentified weaknesses or susceptibilities that predispose them to die suddenly and unexpectedly in infancy. Of course, not all such children die. Another area of agreement is that some kind of stressor is necessary to bring the latent weakness into play, and that's where modifiable parental behaviour and factors such as smoking, sleep position, and co-sleeping enter the picture.

If attempts to identify the "cause" of SIDS have been of little practical use, then by contrast the epidemiological research that has identified several clear triggers has been tremendously successful. In terms of lives saved, this has been money well spent by the foundation on work that is credited with the reduction of SIDS in England and Wales from a peak of 1596 cases in 1988 to fewer than 200 in 2005.[5]

But can the foundation continue to justify funding the hunt for the elusive "cause" of SIDS?

Professor Risdon, until 2006 a member of the foundation's council of trustees, thinks the money invested in such research could be better spent. Professor Haycock, the foundation's scientific adviser, approaches the question this way: "It is absolutely true

that if we were able to apply what we know already, we'd save more lives than we could through laboratory research, and in that sense I think he is right," he says.

"There will always be some babies who die unexpectedly and those parents will need some kind of support. But in terms of justifying expensive research it is getting perhaps quite close to the point where it may be difficult."

In the black and white world of the media, however, the drama of the hunt for a single, silver bullet cause of cot death remains irresistible. Any published research risks being presented as "the answer," rather than being at best just another piece of the puzzle, and there is a danger that such coverage weakens messages about individual parental responsibility.

In November 2006, coverage of the latest "breakthrough" in SIDS research was unequivocal: "Scientists find the key to cot deaths," declared the *Times*,[17] matched by the *Telegraph*'s "Scientists trace 'cause' of cot death to the brain."[18] In fact, David Paterson's team at Boston Children's Hospital had said only that the abnormality of serotonin function they had found in the brainstems of 75% of 31 babies who had died from SIDS—a relatively small number, matched with only 10 controls—"may" be responsible "for a subset of SIDS cases."[19]

A review of world literature published in 1967 identified 11 papers that had indicated that SIDS occurred "more often among the lower socio-economic groups"[20] while as long ago as 1966 one pioneer identified a high prevalence of smoking during pregnancy among mothers whose babies had died suddenly and unexpectedly.[21] Unfortunately, for many years the association with smoking remained a lost observation: three years later, at the second international conference on causes of sudden infant death in infants, at which SIDS was so named, there was not a single reference to the subject.[3]

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Attitudes must change

In the 2000 Confidential Enquiry into Still Births and Deaths in Infancy, Professor Fleming and colleagues called for measures to reduce fetal and infant exposure to tobacco smoke. Smoking in the presence of pregnant women or young infants, they wrote, "should be viewed as being as irresponsible as drinking and driving."[4] Seven years later, they are again calling for legislation to emphasise the adverse effects of tobacco smoke exposure on infants and pregnant women.[9]

Precedents have been set in America, where six states either have, or are considering introducing, bans on smoking in cars in which children are travelling. And last year Arkansas proposed making it an offence for pregnant women to smoke, although it remains unclear how such a law could be enforced.

In the UK, education remains the preferred alternative to legislation, although so far it has failed to reach the 17% of women who smoke throughout their pregnancies[22] and those parents who continue to endanger their infants with other unsafe parenting practices, whether through ignorance or carelessness.

SIDS campaigners, many of them cot death parents whose children died before key risk factors were as well understood as they are today, have worked hard to ensure that parents do not suffer the additional burdens of suspicion and stigma. The flip side of this coin is the need to confront the hard truths about cot death if the "reduce the risk" message is to be brought home to those parents who remain the hardest to reach and whose children are most at risk.[5,23]

Coroners in England and Wales conducting inquests on infants found dead on sofas or in adult beds often give warning of the dangers of co-sleeping, although generally they are careful to avoid any suggestion that the child's death is the parents' responsibility.

Deborah Robinson, an infant death specialist with the SIDS Foundation of Washington, contributed to the Scripps Howard review. In 1991, she lost her own son to SIDS. Today, she trains death scene investigators and believes professionals owe it to parents and children not to varnish the truth.

"Too often I have seen cases where infants die after bed sharing, overlaying, asphyxiation, use of soft bedding, overheating, impairment of caregivers, or exposure to cigarettes and drugs, and so often well meaning professionals, not wanting to further traumatise the family, label it as SIDS," she said.

"Are some of them SIDS? Absolutely, but there are other mechanisms of death involved and yet historically we have thrown all these infant deaths into one category and called them SIDS.

"This topic is very near to my heart on many levels. I am a SIDS mother and understand why professionals do what they do. However, I feel that we do no one justice, especially the infant, if we sugar coat the truths, as hard as they may be."

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